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Case report

Tracheomalatia, to stent or not to stent

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ABSTRACT

Benign thyroid disorders such as goiter, especially retrosternal, can cause tracheostenosis by extrinsic tracheal compression, which is due to the lack of specific symptoms often misdiagnosed. Tracheomalatia develops as a result to long term tracheal compression and refers to weakness of the trachea characterized by softness of the tracheal cartilage arches and by loss of regular tracheal structure. Tracheomalatia is characterized by reduction of the endotracheal lumen and may affect the entire trachea or may be localized to one portion of it. We present the case of a 72-year old patient with distinct tracheostenosis and tracheomalatia, caused by long term pressure by the retrosternal goiter. We have been monitoring the patient for last 20 years after the second endotracheal stent had been placed. The first one was placed 34 years ago, in 1981. On both occasions granulation tissue and colonization of bacteria occurred. In the end the placed stents were rejected and migrated to the main carina. Despite the tracheal diameter narrower than 5 mm the patient has been living normally without the stent for 17 years, with the exception of no hard physical labor. He had a few short term antibiotic therapies and bronchial toilets during symptomatic deteriorations. Diagnosing retrosternal goiter and surgical treatment on time is of crucial importance in cases such as this one. Considering the complications caused by the stent, our opinion is that the majority of patients may require conservative treatment with closely monitoring during respiratory infections.

1. Introduction

Benign thyroid disorders such as goiter can cause tracheostenosis by extrinsic tracheal compression. Substernal goiter is the most frequent cause of thyroid-induced tracheostenosis [1] presenting mostly with the nonspecific respiratory symptoms such as choking, dyspnea and stridor [2] and therefore is often misdiagnosed [3,4]. Long term tracheal compression can cause tracheomalatia which refers to weakness of the trachea. Tracheomalatia is characterized by softness of the supporting tracheal cartilages, extension of the tracheal posterior membranes wall and by reduction of the tracheal antero-posterior diameters [5]. Tracheomalatia can involve the entire trachea or may be localized to one portion of the trachea. Also, it may progress to extensive tracheomalatia, complicated by recurrent and prolonged respiratory infections, episodic choking, hard expectoration and irreversible hypercapnic respiratory failure [6].

2. Case report

We have been monitoring the 72-year old patient with the tracheomalatia for last 20 years. Due to diphtheria in early childhood, he developed tracheal stenosis and by the age of four he had tracheotomy. At the age of 26 years he was hospitalized for surgery of nodular euthyreotic goiter on the both sides which further compressed already stenotic trachea. A partial resection of both lobes of the thyroid gland was performed. At the age of 38 years, after flu, he started having constantly difficult breathing and frequent choking episodes. Tracheoscopy and tracheography confirmed tracheal cicatricial stenosis of 3.5–4 cm in length between the upper and middle third of trachea. Endotracheal lumen was reduced the to 1/3 of normal. A subtotal strumectomy of the

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left lobe of the thyroid gland and an installation of silicone endotracheal (Nevylle) stent were done. Around the stent granuloma formation occurred. Number of respiratory infections caused coughing which bronchiectasis were developed ultimately led to migration of the stent. Stent slipped to the main carina of the trachea, where it remained as a foreign body for 15 years. In the meantime the goiter continued to grow. Scintigraphy and fine-needle aspiration biopsy of the thyroid gland at the age of 53 years showed benign nature nodular changes of both lobes. Due to slippage of the stent, former site of tracheal stenosis became free and was additionally compressed by the growing goiter. Two years later, patient became severely dispnoic, and was frequently burdened with respiratory infections with reduced ability of expectoration. He was hospitalized at the Department of Ear, Nose, Throat, Head and Neck Surgery. MRI and CT of the trachea showed distinctly enlarged right thyroid lobe measuring 9 × 4 cm, filled with well-confined, solid, partly cystic node 4.2 cm in diameter compressing the trachea in length of 4.5 cm from the lower edge of cricoid cartilage to the manubrium of the sternum. At the narrowest point of 2.5 cm of length transversely measured width of the lumen was minimized up to 3.3 mm, and distally from the node was expanded to 3.6 cm. Resection of the right thyroid lobe was performed, and the slipped stent was bronchoscopically pulled out. Surgery intervention proceeded with tracheotomy and implantation of the stent (Rüsch) with metal rings 11 mm in diameter. Due to long term breathing difficulty, bronchiectasis developed. Later that year the patient was frequently hospitalized because of the respiratory infections. He was subjected to prolonged antibiotic therapy according to the antibiogram mainly due to resistant strains of bacteria. Bronchial toilettes with bronchoaspirations were frequent. In 1999, at age of 56 years, despite all precautions, the new stent also slipped from its place. It prevented the patient from breathing and phonating normally. After the tracheoscopy and extraction of the slipped stent, under endotracheal anesthesia, patient's breathing was sufficient and he could phonate normally. Length and place of the tracheostenosis was confirmed with MRI (Fig. 1). Four years later CT showed an extreme tracheal stenosis in 2 cm of length which ended just above the upper edge of manubrium of the sternum (Fig. 2). Frontal diameter at the maximum inspiration was 5 mm and the sagittal diameter 12 mm. Distal from the stenosis trachea had expanded lumen (Fig. 3). That same year

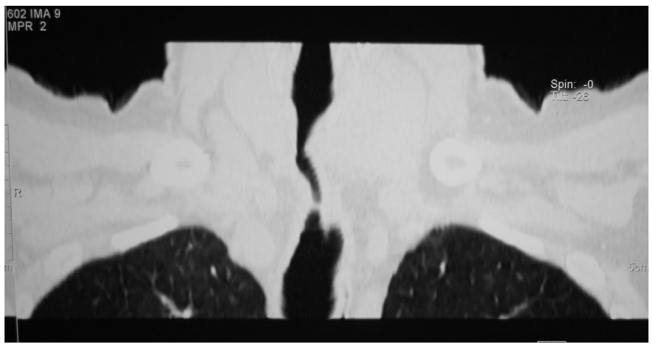

Fig. 2. CT of the trachea performed in 2003 showing extremly tracheostenosis.

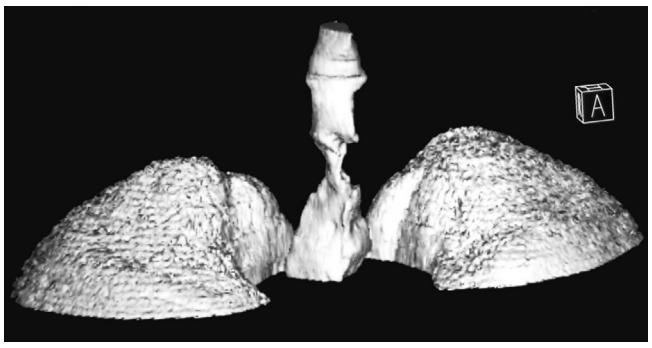

Fig. 3. 3D reconstruction of the trachea performed in 2003 showing stenosis and signs of tracheomalatia distally from the stenosis.

the patient was hospitalized at Department of Internal Medicine due to deteriorated general condition, dyspnea, choking and effort intolerance. Diagnosed was decompensated dilated cardiomiopathy with increased pulmonary artery pressure presumably as a result most likely due to numerous respiratory infections and ischemic genesis. CT performed in 2005 and 2006 showed an enlarged partially resected left thyroid lobe (33 × 46 mm) (Fig. 4) and further narrowing of the sagittal sections of the stenotic tracheal segment to 6 mm. In 2007 the patient was hospitalized

Fig. 1. MRI of the trachea performed in 1999 showing the length and place of the tracheostenosis.

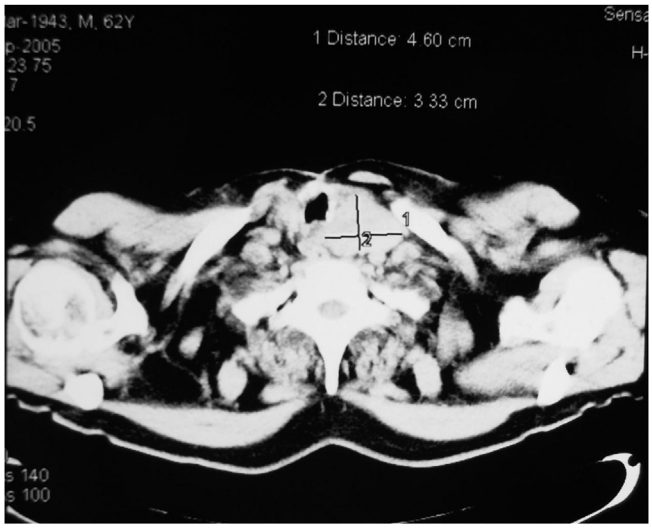

Fig. 4. CT of the trachea performed in 2005 showing enlarged partially resected left thyroid lobe.

again because of symptomatic deterioration at the Department of Pulmonary Diseases. Thread coated with granulation tissue 3 cm below the vocal cords was found during bronchoscopy, and subsequently had been extirpated. Control CT of the chest from the year 2008 showed no changes. In consultation with thoracic surgeon regarding the enlarged left thyroid lobe conservative treatment a conservative approach was selected due to patient's general condition. Since then, the condition of the patient is closely monitored, and there was no need for new interventions.

3. Discussion

In the year 1886 Colles described four cases of tracheostenosis resulting from the need for tracheostomy in diphtheria treatment [7], and Haller was first to describe retrosternal or intratoracic goiter in the year 1749 [2]. After the diagnosis of mediastinal goiter, where 3D CT reconstruction offers optimal accuracy [8], experience indicates that surgical removal of the thyroid gland should be done before the symptoms of compression develop [2,4]. Retrosternal goiter may cause tracheomalatia as a result of pressure on cartilage arches with nodes. In presented case, during the period of 44 years a recurrent retrosternal nodular goiter occurred. As a result of long term pressure deterioration of cartilage arches and walls of the trachea occurred causing a significant narrowing of the tracheal lumen. The endotracheal stent was twice implanted without long term success. On both occasions, up to 2 years after implantation, the stent migrated on carina of trachea. Some experts state that the silicone stent is an option in the treatment of the tracheal stenosis [9], but the metal stent is considered the most effective therapy for tracheostenosis which appears as a result of nodal goiter [3,10,11]. Both have their advantages and disadvantages, and their effectiveness is individual, as in our case. Metal stent after implantation is difficult to remove because it is inserted into the bronchial mucosa and epithelializate inside. It is associated with the development of many complications, and therefore requires bronchoscopic surveillance and often further therapeutic interventions [10,12]. Possible complications after stent implantation are growth of the granulation tissue in the lumen of the trachea and displacement of the prosthesis [10,13,14] as was in our case. Silicone stent can lead to retention of the bronchial secretion, and also migration and granulation [14]. In case of above mentioned complications removal of the stent is required, as was done in our case. Both times after removal of the stent the patient continued with his everyday life without physical strains. Despite narrowing of the frontal endotracheal lumen to the diameter of 3.3 mm, the patient tolerates well moderate physical activity and usual daily activities. He is regularly monitored and, if necessary, a bronchial toilette with bronchoaspiration is performed. According to the results of microbiologic sputum analysis or the bronchial aspirate antibiotic therapy is administered. Current experience indicates that the total thyroidectomy is treatment of choice, not the subtotal thyroidectomy as it was done in presented case [2,4]. The question is whether the implantation of the stents were even necessary and whether their implantation led to further infections deteriorating fibrotic changes and granulation in the tracheal mucosa.

4. Conclusion

With intensive examination of the first symptoms such as the pressure behind the sternum, retrosternal goiter had to be diagnosed as early as possible followed by surgery, in this case total thyroidectomy. Stenting in this case was not effective. Considering the complications caused by the stent, long-term survival in presented patient and good quality of life even with narrowing of the endotracheal lumen below the diameter of 5 mm, our opinion is that the majority of patients may require conservative treatment with closely monitoring during respiratory infections.

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